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Stigmatization predicts psychological adjustment and quality of life in children and adolescents with a facial difference

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Abstract: OBJECTIVES: This cross-sectional study assessed psychological adjustment and health-related quality of life (HRQOL) in children and adolescents with congenital or acquired facial differences and identified potential predictors of adjustment. METHODS: Data were obtained from 88 children, ages 9 months to 16 years, by means of parent questionnaires (n = 86) and standardized interviews with children 7 years old (n = 31). Evaluation measures included the Child Behavior Checklist (CBCL), KIDSCREEN-27, TNO-AZL Preschool Quality of Life Questionnaire (TAPQOL), and Perceived Stigmatization Questionnaire. RESULTS: Psychological adjustment, as measured by the CBCL, was within norms. Parent-reported HRQOL was good in preschool children. Parent- and self-reported HRQOL of participants 7-16 years old was impaired in several dimensions, including psychological well-being. Psychological adjustment (especially internalizing behavior problems) and HRQOL were predicted primarily by perceived stigmatization. CONCLUSIONS: Identification of stigma experiences and appropriate support may be crucial to enhancing psychological adjustment and quality of life in children with facial disfigurement.

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Stigmatization Predicts Psychological Adjustment and Quality of Life in Children and Adolescents With a Facial Difference

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Objectives This cross-sectional study assessed psychological adjustment and health-related quality of life (HRQOL) in children and adolescents with congenital or acquired facial differences and identified potential predictors of adjustment. **Methods** Data were obtained from 88 children, ages 9 months to 16 years, by means of parent questionnaires ($n = 86$) and standardized interviews with children ≥ 7 years old ($n = 31$). Evaluation measures included the Child Behavior Checklist (CBCL), KIDSCREEN-27, TNO-AZL Preschool Quality of Life Questionnaire (TAPQOL), and Perceived Stigmatization Questionnaire. **Results** Psychological adjustment, as measured by the CBCL, was within norms. Parent-reported HRQOL was good in preschool children. Parent- and self-reported HRQOL of participants 7–16 years old was impaired in several dimensions, including psychological well-being. Psychological adjustment (especially internalizing behavior problems) and HRQOL were predicted primarily by perceived stigmatization. **Conclusions** Identification of stigma experiences and appropriate support may be crucial to enhancing psychological adjustment and quality of life in children with facial disfigurement.

Facial differences can result from a wide range of conditions, including congenital malformations (e.g., cleft lip, port wine stains, nevi), injuries (e.g., burns), and dermatological diseases (e.g., psoriasis). Despite important advances in medical and surgical interventions, complete resolution of such conditions is rarely obtainable. Therefore, it is important to examine the consequences of a facial difference on the psychological adjustment and health-related quality of life (HRQOL) of affected individuals.

Facial appearance exerts a strong impact on social interaction and personal development (Langlois et al., 2000). Consequently, facial differences are presumed to negatively affect social encounters and to put individuals

at risk for psychosocial difficulties and impaired HRQOL (Topolski, Edwards, & Patrick, 2005). Research findings confirm that individuals with visible differences are likely to experience stigmatizing behaviors, such as staring, avoiding, teasing, and manifestations of pity (Lawrence, Rosenberg, Mason, & Fauerbach, 2011; Masnari et al., in press; Strauss et al., 2007). Yet, data on the psychological adjustment and HRQOL of children and adolescents with visible differences are controversial. Several studies among individuals with various facial conditions have reported no major psychological maladjustment (Dieterich-Miller, 1992; Landolt, Grubenmann, & Meuli, 2000; Sheerin, MacLeod, & Kusumakar, 1995). However, there is some evidence of impaired HRQOL and difficulties in particular

areas of functioning, the most frequent of which relate to negative self-perceptions, emotional problems, and social functioning (Hunt, Burden, Hepper, & Johnston, 2005; Stubbs et al., 2011; Topolski et al., 2005).

Previously suggested determinants of adjustment to visible differences in children include medical variables, characteristics of the child, and family/social variables. There is some evidence that acquired conditions are associated with more difficulties than congenital conditions (Patrick et al., 2007). It has been suggested that condition-specific effects are less important predictors of individual adjustment than one might expect (Rumsey and Harcourt 2007). Notably, numerous studies have shown that the severity of a condition is not a reliable predictor of psychological distress (Thompson & Kent, 2001). Findings about the impact of socioeconomic status (SES), age, and gender are inconsistent (Hunt et al., 2005). The quality of family relationships, parental adjustment, and social support have been found to be of great importance (Noronha & Faust, 2007). Teasing and bullying experiences have been shown to be negatively associated with mental health (Hunt, Burden, Hepper, Stevenson, & Johnston, 2007; Rimmer et al., 2007). Yet, to our knowledge, there is no quantitative evidence on the association between child adjustment and perceived stigmatization, which includes not only experiences of teasing, but also exposure to staring and other disrespectful behaviors.

Current data on psychological adjustment and HRQOL in children and adolescents with facial differences are limited in several ways. First, studies on appearance-altering conditions (e.g., burn scars, infantile hemangioma, and congenital melanocytic nevi) seldom differentiate between subjects with facial and nonfacial differences, thereby making specific conclusions difficult. Second, most of the existing research on facial disfigurement has focused on subjects with a cleft lip/palate or other severe craniofacial malformations. These conditions often involve functional impairment (e.g., speech or eating difficulties); thus, findings may not be generalized to conditions that cause only esthetic impairment (e.g., port wine stains). Finally, little data exist on predictors of adjustment specific to facial differences, and there is no quantitative evidence on the impact of perceived stigmatization.

The objectives of our study were twofold. First, we aimed to assess psychological adjustment and HRQOL in young people with facial differences exerting an esthetic but no functional impact. We expected to find good overall psychological adjustment and good overall HRQOL, albeit with some impairment in the social and emotional domains. Second, we aimed to examine the importance of medical, individual, and family-related predictors of

adjustment to facial differences. Based on previous findings, we expected the size of the facial difference not to be predictive of adjustment. In contrast, we expected perceived stigmatization to be a significant predictor of child psychological adjustment and HRQOL.

Methods

Participants and Procedure

The data presented in this article are part of comprehensive data collection assessing the psychosocial impact of facial differences in children and adolescents. Data on the frequency of child- and parent-reported stigma experiences as well as predictors of perceived stigmatization are presented elsewhere (Masnari et al., 2012).

Participants were recruited among outpatients of University Children's Hospital Zurich, Switzerland, and University Medical Hospital Freiburg, Germany. The study was approved by the local ethics committee. Families were eligible for the study if their child met the following criteria: (a) a visible facial difference (burn scar, infantile hemangioma, port wine stain, or congenital melanocytic nevus) with a current size $\geq 1 \text{ cm}^2$; (b) age between 9 months and 16 years; (c) at least 6 months postaccident for burn patients; (d) no evidence of mental retardation; and (e) a good understanding of German. Based on the medical records of the two hospitals, 126 eligible families were identified and contacted by letter: nine could not be reached, 15 did not respond, 11 refused participation, and 3 were excluded from analyses owing to incomplete data. Thus, 88 families were included (response rate = 69.8%). Nonparticipants consisted mainly of children with infantile hemangiomas ($n = 28$) and were slightly younger than participants ($M_{\text{age}} = 4.54$ vs. 6.31 years; $t = -2.37$, $p < .05$).

Data were obtained by means of parent questionnaires and standardized interviews with children aged ≥ 7 years. Parent-provided proxy reports on their child's psychological well-being and HRQOL as well as information on possible predictors, including sociodemographic and medical variables as well as self-reports on their own mental health. Children ≥ 7 years old provided self-reports of their HRQOL.

Parents provided written informed consent. Parents of children < 7 years old received standardized questionnaires by mail. Parents of children ≥ 7 years old were asked to return an answer form indicating whether they agreed to participate in the study and whether their child was willing to take part in a face-to-face interview. These standardized interviews were conducted by the first author either at the child's home or at the hospital. To ensure that children

could express their own views openly, they were interviewed separately from their parents. Parent questionnaires were handed out after the child interview. These included three separate booklets: one asked parents to conjointly provide information on their child (proxy ratings); the two other booklets asked mothers and fathers to provide information separately on their own mental health. In two of the 34 cases involving a child ≥ 7 years old, only the child participated, whereas in three of these cases, only the parent responded. Consequently, 54 parent reports were obtained for preschool children, and 32 parent and 31 self-reports for school-age children. In 35 cases, proxy ratings were provided by the mother, in seven cases by the father, and in 37 cases by both parents conjointly. In seven cases, which parent filled out the proxy form was not indicated. If families did not respond to the initial study invitation or if questionnaires were not returned within 2 weeks, a reminder was sent by mail. After two subsequent weeks without notice, families were contacted by phone. There was no remuneration for participating in the study; but travel costs were reimbursed.

Measures

Health-Related Quality of Life

HRQOL for children ages 9 months to 6 years was assessed by parental report, using an authorized German version of the TNO-AZL Preschool Quality of Life Questionnaire (TAPQOL) (Fekkes, Bruil, & Vogels, 2004). HRQOL for participants ages 7–16 years was assessed using the German parent and child form of the KIDSCREEN-27 (Bisegger, Cloetta, & the European KIDSCREEN Group, 2005).

The TAPQOL is a well-validated, standardized and reliable measure that assesses proxy reports of HRQOL in preschool children. It consists of 43 items, classified into 12 syndrome scales, assessing four global dimensions of HRQOL: physical, social, cognitive, and emotional functioning. Three syndrome scales (social functioning, motor functioning, and communication) are applicable only to children ≥ 1.5 years of age. Following the TAPQOL protocol, parents were asked to note problems in any of the mentioned domains (e.g., *Has your child had stomachache or abdominal pain?*) and to rate their child's well-being related to the specific problem (*At that time, my child felt fine, not so good, quite bad, bad*). All items had a recall period of 1 week. A description of the items can be found in Fekkes et al. (2000). Syndrome scales were transformed into a 0–100 scale. Higher scores indicate better HRQOL. To obtain a measure of overall HRQOL, we computed a total score by averaging the scores of the four global dimensions, which previously were computed

as the average of all underlying syndrome scales. Norms were retrieved from the scale manual and were based on data from 251 parents of healthy Dutch children between the ages of 10 and 60 months (Fekkes et al., 2004). Internal consistency in this study was acceptable to good for the total score and most syndrome scales, except for the scales measuring lung, stomach, social, and anxiety problems, which revealed poor internal consistencies (Table II).

The KIDSCREEN-27 is a standardized multidimensional generic instrument designed to assess self- and proxy-reported HRQOL in children and adolescents 8–18 years old. The parent form was administered as a questionnaire and the child form as an interview. Validity and reliability of this instrument have been confirmed (Ravens-Sieberer et al., 2007). The questionnaire contains 27 items assessing five dimensions: physical well-being, psychological well-being, parent relations and autonomy, social support and peers, and school environment. The items assess either the frequency or the intensity of a behavior or a feeling on a 5-point Likert scale, over a recall period of 1 week. Following the Swiss manual (Bisegger et al., 2005), scale scores were transformed into *T* values based on reference data from a community sample of >1,600 Swiss children and parents. In our study, children 7 years old ($n = 3$) were thereby compared with normative data of 8–11 year olds. A total score was computed by averaging the *T* scores over the five global scales. Internal consistency in this study was acceptable to good for the total score and most subscales for both self- and proxy reports, except for the subscale “school environment” (Table II).

Psychological Adjustment

The Child Behavior Checklist (CBCL) is a widely used, well-validated, standardized measure assessing parental reports of a child's psychological adjustment (Achenbach, 1991; Achenbach & Rescorla, 2000). Two authorized German versions of the CBCL were used: the CBCL/1.5-5 (Arbeitsgruppe Deutsche Child Behavior Checklist, 2002) for children ages 18 months to 4 years and the CBCL/4-18 (Steinhausen, Winkler Metzke, & Kannenberg, 1996) for children ages 4–16 years. Both instruments yield scores for two broadband scales (internalizing and externalizing behavior problems), and an overall total behavioral problems score. Higher scores indicate greater psychological maladjustment. *T* scores were derived based on normative data. For the CBCL/4-18, reference values were drawn from 1964 healthy Swiss children (Steinhausen et al., 1996). For the CBCL/1.5-5, no Swiss/German norms are available. Therefore, *T* scores were calculated based on a community sample of 700 healthy U.S. children (Achenbach & Rescorla, 2000). In the current study,

internal consistency for the internalizing ($\alpha = .73/.87$), externalizing ($\alpha = .90/.93$), and total behavioral problems scales ($\alpha = .92/.95$) was acceptable to excellent for both the CBCL/1.5-5 and the CBCL/4-18, respectively.

Perceived Stigmatization

Child stigma experiences were assessed via a German translation of the parent form of the Perceived Stigmatization Questionnaire (PSQ) (Lawrence, Rosenberg, Rimmer, Thombs, & Fauerbach, 2010). The translation procedure followed published guidelines (Brislin, Lonner, & Thorndike, 1973), including the use of independent back-translation. The parent form of the PSQ asks parents to rate how often their child experienced a variety of stigmatizing behaviors commonly reported by people with appearance distinctions. It contains 21 items classified into three factors: absence of friendly behavior, staring/confused behavior, and hostile behavior. Answer choices are on a 5-point Likert scale, ranging from 1 (*never*) to 5 (*always*), with a recall period of 1 year. A PSQ total score is obtained by averaging over all items. Higher scores indicate higher perceived stigmatization. A recent study confirmed the good psychometric properties of this instrument (Lawrence et al., 2010). In the current study, internal consistency for the PSQ total score was good for both self- ($\alpha = .81$) and proxy reports ($\alpha = .88$).

Size of the Facial Difference

As many participants in our study were outpatients not regularly seen by a physician, we had no up-to-date medical records for all of them. Therefore, we assessed the size of the facial difference by parent estimate. Parents were asked to draw the extent of their child's facial difference on a face template. In the two cases in which only the child participated, this information was assessed by the first author at the interview with the child. The size of the facial difference was categorized into four groups, according to the extent of the face affected by the condition: ≤ 5 , $>5-25$, $>25-50$, or $>50\%$. The initial categorization was performed by the first author who carried out the interviews with the patients. The second author assessed this classification for 20 randomly selected participants. With agreement of 95%, inter-rater reliability was excellent.

Mental Health of Parents

Mental health of mothers and fathers was assessed independently with the Symptom Checklist-27 (SCL-27), a well-validated multidimensional measure (Hardt, Egle, Kappis, Hessel, & Brahler, 2004). The Global Severity Index (GSI) was used as an indicator of mental health. Higher scores indicate poorer mental health. The SCL-27

was filled out by 85 mothers and 78 fathers; in 78 cases, data were available from both parents. Internal consistency of the GSI was $\alpha = .89$ for mothers and $\alpha = .70$ for fathers.

Socioeconomic Status

SES was calculated as a sum score (range: 2–12) based on paternal occupation and maternal education. Specific examples of occupational and educational levels were provided in a previous article (Largo, Molinari, Comenale, Weber, & Duc, 1989). Occupational levels were assessed on a 6-point scale ranging from 1 (*occupations that do not require any school qualifications or vocational training*) to 6 (*occupations that require a University degree*). Accordingly, education was categorized into six levels from 1 (*did not graduate from compulsory school*) to 6 (*University degree*). For mothers, level of education was used instead of occupation because, in Switzerland, mothers of young children often resign from their jobs after their child's birth to stay at home with their children. Three social classes were defined as follows: scores 2–5 as lower SES, scores 6–9 as middle SES, and scores 10–12 as upper SES. For statistical analysis, we used the sum score. A similar measure has been used before and has been shown to be a valid measure of SES in the Swiss community (Landolt, Buehlmann, Maag, & Schiestl, 2009).

Statistical Analyses

Data were analyzed using the statistical package PAWS for Windows, release 18. All analyses were performed with two-tailed tests and $p < .05$ considered significant. For categorical comparisons, we used χ^2 -tests. For comparisons of continuous data, we used Student's *t*-tests. The differences between sample means and reference data were quantified by calculating effect sizes (Cohen's *d*; 0.2 small, 0.5 medium, 0.8 large effect size) (Cohen, 1988). To obtain a comparable measure of parent-reported HRQOL for all participants, the parent-reported TAPQOL and KIDSCREEN-27 total scores were both transformed into norm-based *t* scores and combined into a single variable: the parent-reported HRQOL total score. Four linear regression models were generated using the parent-reported HRQOL total score and the three CBCL scores (total, internal, and external behavioral problems score) as dependent variables, all of which were normally distributed. The predictors were entered hierarchically in blocks; within blocks, variables were entered simultaneously. The four variable blocks are as follows: (1) child age, child gender, SES; (2) maternal and paternal GSI; (3) type (*acquired* vs. *congenital*) and size of facial difference; and (4) perceived stigmatization. We chose this method to ensure that the effect of the medical variables and the perceived

stigmatization were controlled for the variance contributed by sociodemographic variables and parental mental health. Owing to the small sample size in self-reports ($n = 31$), we elected not to attempt regression analysis for self-reported HRQOL.

Results

Sample Characteristics

Sample characteristics are summarized in Table I for the overall sample, as well as for preschool and school-age children separately. The majority of the preschool children had a congenital condition, whereas almost 60% of the school-age children had a burn scar. Also, small facial differences, covering 5% or less of the face, were more frequent in the younger age group. Notably, in general, most of the patients included in this study had a facial difference affecting the skin only, without marked distortion of facial features and without functional impairment. Parent-perceived stigmatization of their child was significantly higher in the older age group. The mental health of parents and SES of families did not differ between the two age groups. Most families were from the middle or upper class.

Psychological Adjustment

Psychological adjustment, as measured by the CBCL, was well within norms. Parents in our sample ($n = 72$) reported their children to have no more internalizing ($M = 49.32$, $SD = 10.45$, $p = .58$, $d = .07$), externalizing ($M = 49.82$, $SD = 10.92$, $p = .89$, $d = 0.02$), or total behavior problems ($M = 50.07$, $SD = 11.03$, $p = .96$, $d = 0.01$) than a community sample.

Health-Related Quality of Life

Tables II and III show the mean scores of the HRQOL measures for our sample and the reference groups. Parents of children ages 9 months to 6 years did not report any impairment of their child's HRQOL as measured by the TAPQOL. Indeed, they described their child as more active (lively/energetic) and having a better appetite than the reference group. Conversely, overall parent-reported HRQOL for patients 7–16 years old was impaired relative to community norms. Specifically, physical, psychological, and school functioning were significantly poorer, exhibiting small to moderate effect sizes. Parent relations and autonomy also were slightly impaired, albeit not significantly. Social support, however, was reported to be normal. Self-reports of HRQOL were within published norms, except for one dimension: children and adolescents with a facial difference reported poorer psychological well-being. Notably, social support was slightly better

than in the community sample, although this effect did not reach the significance level.

Predictors of Psychological Adjustment and HRQOL

Table IV summarizes statistics for the four regression models predicting proxy-reported psychological adjustment and HRQOL. The selected predictors accounted for 24% of the variance in the CBCL total behavior problem score, 34% of the internalizing and 8% of the externalizing behavior problem score, and 32% of the HRQOL score. All models were statistically significant, except for the one predicting externalizing behavior problems ($p = .14$). Child age and gender did not significantly predict either outcome variable. SES status was a significant predictor of HRQOL. With the entry of the parental mental health indexes in Step 2, there was a significant increase of the amount of variance explained for all outcome variables. However, maternal and paternal mental health scores, separately, were not significant predictors of the dependent variables, except for the internalizing behavior problem score, which was significantly predicted by paternal mental health. Notably, medical variables (i.e., the type (*acquired* vs. *congenital*) and size of the facial difference), entered in Step 3, did not significantly predict any outcome variable. Conversely, perceived stigmatization, entered in Step 4, accounted for a significant portion of the variance in all outcome variables, except for the externalizing behavior problem score. Thus, children and adolescents experiencing high levels of stigmatization were at greatest risk of psychological maladjustment (especially internalizing behavior problems) and low HRQOL.

Discussion

The purposes of this study were to assess psychological adjustment and HRQOL in children and adolescents with various kinds of facial difference and to identify possible predictors of adjustment. In line with our first hypothesis, parents in our sample reported no significant psychological maladjustment of their child as measured with the CBCL. This result is consistent with previous findings in children with facial hemangiomas, burns, or port wine stains (Dieterich-Miller, 1992; Landolt et al., 2000; Sheerin et al., 1995). Although one could argue that generic measures are not able to capture the specific problems of individuals with facial differences, this result still suggests that the children in our sample might not suffer from any major psychological maladjustment.

Table I. *Sample Characteristics*

Characteristic	Total sample (<i>n</i> = 88)	Preschool children (<i>n</i> = 54)	School-age children (<i>n</i> = 34)	Comparison of the two age groups		
				χ^2	<i>t</i>	<i>p</i>
Age (year), <i>M</i> (<i>SD</i>)	6.31 (4.66)	3.08 (1.80)	11.44 (2.80)		15.48	<.001
Range (year)	0.75–15.75	0.75–6.67	7.00–15.75			
Gender				3.84		.08
Female	40 (45.5)	29 (53.7)	11 (32.4)			
Male	48 (54.5)	25 (46.3)	23 (67.6)			
Type of condition						
Burn scar	25 (28.4)	5 (9.3)	20 (58.8)	25.2		<.001
Port wine stain	19 (21.6)	10 (18.5)	9 (26.5)	.78		.43
Infantile hemangioma	36 (40.9)	31 (42.6)	6 (17.6)	13.54		<.001
Congenital melanocytic nevus	8 (9.1)	8 (14.8)	0	5.54		.02
Size of facial difference						
≤5% of the face	28 (31.8)	25 (46.3)	3 (8.8)	13.50		<.001
>5–25% of the face	37 (42.0)	19 (35.2)	18 (52.9)	2.70		.12
>25–50% of the face	15 (17)	6 (11.1)	9 (26.5)	3.48		.08
>50% of the face	8 (9.1)	4 (7.4)	4 (11.8)	.48		.71
PSQ total score, <i>M</i> (<i>SD</i>) ^a	1.82 (0.49)	1.66 (0.40)	2.10 (0.53)		3.82	<.001
Parental mental health						
GSI mother, <i>M</i> (<i>SD</i>) ^b	0.32 (0.36)	0.31 (0.37)	0.33 (0.33)		0.24	.81
GSI father, <i>M</i> (<i>SD</i>) ^c	0.24 (0.24)	0.26 (0.25)	0.20 (0.22)		−0.95	.35
Socioeconomic status, <i>M</i> (<i>SD</i>)	9.07 (2.02)	9.25 (1.82)	8.77 (2.33)		−1.03	.31
Lower	3 (3.4)	1 (1.9)	2 (5.9)			
Middle	45 (51.1)	27 (50.0)	18 (52.9)			
Upper	36 (40.9)	25 (46.3)	11 (32.4)			
Unknown	4 (4.5)	1 (1.9)	3 (8.8)			

Note. Frequencies are reported in absolute numbers of cases and percentages in parentheses.

Sample sizes vary due to missing data: ^a*n* = 84 (53/31), ^b*n* = 85 (53/32), ^c*n* = 78 (49/29).

With regard to HRQOL, our hypothesis that children with facial differences would fare worse than controls was supported for school-aged children, but not for preschool children. In preschool children, parent-reports of child HRQOL were well within norms. This result is in contrast with previous findings among children with facial burns (Stubbs et al., 2011) or infantile hemangiomas (Hoornweg, Grootenhuis, & van der Horst, 2009). Several factors may explain this discrepancy, including differences in the measures used or certain characteristics of the samples. Stubbs et al. (2011), for example, assessed HRQOL with a burn-specific questionnaire, which is possibly more sensitive to appearance-related difficulties than our questionnaire. Hoornweg et al. (2009) used the same measure as in our study, but 34% of the children in their sample had hemangioma-related complications (e.g., bleeding or impaired vision), which was not the case in our sample. This may explain the better HRQOL reports in our study. For patients ages 7–16 years, parents reported a significantly poorer overall HRQOL compared with reference data. Specifically, physical, psychological, and school

functioning were impaired, with small to moderate effect sizes. With regard to self-reported HRQOL, children and adolescents in our sample exhibited good overall HRQOL, but diminished psychological well-being. Findings of impaired HRQOL in this age group are in line with previous research among individuals with different facial conditions (Stubbs et al., 2011; Topolski et al., 2005). Interestingly, in our sample, social support was normal in both self- and parent reports. This supports the observation that, although a facial difference may cause some difficulties in social encounters, it does not prevent children from having close friendships (Feragen, Kvale, Rumsey, & Borge, 2010).

The different outcomes in the two age groups may be related to a number of factors. First, the two HRQOL measures used in this study could have assessed different aspects of quality of life. Second, different reference groups were used for the two age groups; this may have influenced the evaluation of outcomes. Third, the two subsamples differed regarding the type and size of the participants' facial differences. However, as multivariate analysis

Table II. Sample Means and Reference Data for Health-Related Quality of Life in Preschool Children

Measure	Sample			Reference group ^a		Effect size	
	<i>n</i>	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>	<i>d</i>	<i>p</i> ^b
TAPQOL parent form							
Physical functioning							
Sleeping ($\alpha = .86$)	54	82.21	18.51	83.10	16.84	0.05	.74
Appetite ($\alpha = .74$)	54	94.29	8.99	85.93	12.27	0.78	<.001
Lung problems ($\alpha = .51$)	54	94.75	11.13	97.24	8.52	0.25	.13
Stomach problems ($\alpha = .48$)	54	91.82	13.10	92.60	13.23	0.06	.70
Skin problems ($\alpha = .81$)	54	91.20	13.00	92.83	10.08	0.14	.31
Motor functioning ($\alpha = .67$)	40	98.28	5.29	98.54	4.23	0.05	.73
Social functioning							
Social functioning ($\alpha = .58$)	40	94.58	11.56	91.43	15.00	0.24	.14
Problem behavior ($\alpha = .90$)	53	73.32	23.90	67.75	15.38	0.28	.11
Cognitive functioning							
Communication ($\alpha = .82$)	39	91.83	12.09	91.69	9.92	0.01	.94
Emotional functioning							
Anxiety ($\alpha = .51$)	52	83.65	17.30	79.22	17.85	0.25	.10
Positive mood ($\alpha = .80$)	53	97.80	8.67	98.94	5.67	0.16	.36
Liveliness ($\alpha = 1.00$)	53	100.00	0.00	98.07	7.74	0.35	<.001
Total score ($\alpha = .71$)	53	89.29	7.95	88.13	5.99	0.16	.32

Note. The scales "motor functioning," "social functioning," and "communication" are only relevant for children aged ≥ 18 months.

α = Cronbach alpha.

^aThe reference group consists of 251 parents of healthy Dutch children between the ages of 10 and 60 months (Fekkes et al., 2004)

^bTwo-sample *t*-test with normative data.

Table III. Sample Means and Reference Data for Health-Related Quality of Life in School-Age Children

Measure	Sample			Reference group ^a		Effect size	
	<i>n</i>	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>	<i>d</i>	<i>p</i> ^b
KIDSCREEN-27 parent form							
Physical well-being ($\alpha = .92$)	32	46.56	15.72	52.81	8.69	0.49	.03
Psychological well-being ($\alpha = .88$)	32	45.85	12.21	51.90	8.98	0.56	.01
Autonomy & parents ($\alpha = .69$)	32	49.41	11.46	53.14	8.36	0.37	.08
Social support ($\alpha = .88$)	32	50.25	12.66	51.04	8.12	0.07	.73
School environment ($\alpha = .49$)	31	49.01	6.60	52.52	8.92	0.45	.01
Total score ($\alpha = .86$)	31	48.75	9.21	52.28	8.61	0.40	.04
KIDSCREEN-27 child form							
Physical well-being ($\alpha = .86$)	31	51.16	12.45	52.66	9.00	0.14	.51
Psychological well-being ($\alpha = .78$)	31	49.07	9.93	53.05	9.26	0.41	.03
Autonomy and parents ($\alpha = .66$)	31	54.68	11.23	53.38	8.77	0.13	.53
Social support ($\alpha = .69$)	31	54.00	8.99	50.98	8.97	0.34	.07
School environment ($\alpha = .44$)	31	53.50	6.84	52.95	9.02	0.07	.66
Total score ($\alpha = .65$)	31	52.48	6.48	52.60	9.00	0.02	.92

Note. α = Cronbach alpha.

^aThe reference group consists of >1,600 Swiss children and parents (Bisegger et al., 2005).

^bOne-sample *t*-test with normative data.

demonstrated that these variables were not predictive of HRQOL, this seems not to be a decisive factor. Finally, it could be speculated that the psychosocial impact of a facial difference increases with age. Our data suggest that school-age children are more at risk for experiencing

social stigmatization than younger children. As several parents in our sample have commented, toddlers may be too young to be aware of their condition. Yet, problems may evolve in early school years when children increasingly engage in social comparisons with peers (Rumsey &

Table IV. Predictors of Parent-Reported Psychological Maladjustment and Health-Related Quality of Life

Predictor	Parent reported psychological maladjustment (<i>n</i> = 64)						Parent-reported health-related quality of life (<i>n</i> = 75)	
	Total behavior problems		Internalizing behavior problems		Externalizing behavior problems			
	ΔR^2	β	ΔR^2	β	ΔR^2	β	ΔR^2	β
Step 1	.05		.13*		.03		.10*	
Child age		−.06		.09		−.12		.10
Gender of child		.19		.12		.22		−.08
Socioeconomic status		−.01		−.17		−.03		.25*
Step 2	.16**		.19**		.10*		.09**	
Mental health (GSI) of mother		.16		.16		.09		−.18
Mental health (GSI) of father		.16		.26*		.18		−.07
Step 3	.04		.01		.03		.01	
Acquired vs. congenital condition		.25		.18		.23		−.17
Size of facial difference		.08		−.03		.06		.21
Step 4	.09**		.09**		.04		.19***	
Parent-perceived stigmatization		.36**		.35*		.23		−.53***
Total <i>R</i> ²	.34**		.42**		.19		.40***	
Total <i>R</i> ² adjusted	.24**		.34**		.08		.32***	

Note. * $p < .05$, ** $p < .01$, *** $p < .001$

Harcourt, 2007). Problems also may arise in adolescence, when appearance, peer approval, and identity issues become paramount (Edwards et al., 2005). In addition, adolescents may increasingly have to deal with difficulties by themselves, whereas younger children may benefit from greater parental support. All this is likely to cause greater vulnerability in older children and adolescents than in pre-school children. On the flip side, age also may confer adjustment benefits through the development of coping strategies over time (Thompson & Kent, 2001). Our data do not allow any conclusive explanation for the different outcomes in the two age groups; this issue requires further longitudinal research.

Multivariate analysis revealed that psychological adjustment and HRQOL were not predicted by child age and gender. But good HRQOL was associated with high SES, which is in line with previous findings (Bradley & Corwyn, 2002). The simultaneous entry of maternal and paternal mental health indexes into the regression model contributed significantly to the prediction of all outcome variables. However, maternal and paternal mental health as individual variables, were not significant predictors for the outcome variables, except that paternal mental health significantly predicted child internalizing behavior problems. In line with our hypothesis and previous findings (Thompson & Kent, 2001), the size and the type (*congenital* vs. *acquired*) of the facial difference did not significantly predict psychological adjustment or HRQOL. This supports the notion that all children who look different face similar problems, irrespective of their specific condition

(Rumsey & Harcourt, 2007). Notably, global psychological adjustment and HRQOL were primarily predicted by perceived stigmatization. Particularly, perceived stigmatization predicted significantly internalizing, but not externalizing behavior problems. This fits well with the notion that children with visible differences are especially at risk for internalizing problems, like anxiety, depression, and social withdrawal. To date, literature concerning the association between stigmatization and adjustment to disfigurement has been predominantly of a theoretical nature and based on qualitative research. This is the first study to provide quantitative evidence on this association in young people with facial differences. Concordant findings have been documented in research among adults with psoriasis (Richards, Fortune, Griffiths, & Main, 2001). The strong association between stigmatization and adjustment may be explained by several mechanisms. First, through stigma experiences, individuals with a facial difference could conclude that they are deficient relative to their peers, which can lead to negative self-images and subsequent psychological difficulties (Hunt et al., 2007). Second, affected individuals could react with avoidance of potentially painful social encounters, which again, may constrain their psychosocial development (Kish & Lansdown, 2000). Third, teasing and poor peer acceptance may contribute to emotional problems, like an increased sense of loneliness and social isolation (Rumsey & Harcourt, 2007).

The strengths of the current study are its inclusion of children with a broad spectrum of ages and facial

differences, high participation rate, use of well-validated, multidimensional and standardized measures with reference data, assessment of self- and proxy reports, inclusion of fathers, and adoption of multivariate statistics. Post hoc power analysis ($\alpha = .05$, two-tailed) using the G*power software (Faul, Erdfelder, Lang, & Buchner, 2007) indicated that for each *t*-tests comparing sample means with normative data, the power to detect a large effect size ($d = 0.8$) exceeded .99. The power to detect a medium effect size ($d = 0.5$) was adequate for the CBCL (.99) and the TAPQOL (.83), but just below the recommended .80 level (Cohen, 1988) for the KIDSCREEN-27 (.77). The power to detect a small effect size ($d = 0.2$) was less than adequate for all outcome variables. For the regression analyses, the power to detect a large ($f^2 = 0.35$) or a medium ($f^2 = 0.15$) effect size was more than adequate (i.e., $>.80$) in each analysis, but the power to detect small-sized effects ($f^2 = 0.02$) was less than adequate. Thus, overall, our sample sizes provided adequate power at the medium to large effect size level, but not enough power at the small effect size level.

Further limitations to the present study exist. First, the cross-sectional design of this study prevents any conclusions about causal relations. Second, the exclusion of non-German-speaking families resulted in an underrepresentation of lower class families. Third, the appropriateness of using U.S. norms for the CBCL/1.5-5 and Dutch norms for the TAPQOL can be questioned. Slight differences in the age ranges of the normative samples and cultural differences may compromise comparability. This being said, cross-cultural bias seems unlikely, given previous findings supporting the use of U.S. norms for the CBCL in German samples (Elting, 2003), as well as similar HRQOL scores of chronically ill children within central European countries (Schmidt et al., 2006). Fourth, some subscales of the HRQOL measures exhibited low internal consistency. This raises doubts on the usefulness of these subscales in future research and calls for a revaluation of their validity. However, the low internal consistency of some subscales did not compromise our multivariate analyses, for which we only used global HRQOL scores with good internal consistency. Fifth, the size of the facial difference was estimated based on parent perception, though categorized by raters; future research should try to implement a standardized measurement. Sixth, in multivariate analyses the use of both parent-reported predictors and outcome variables did not control for common source and method variance as factors that might contribute to significant findings. Finally, we elected not to attempt multivariate analysis with self-reported HRQOL, because of the small

sample size for self-reports ($n = 31$); this issue warrants further investigation.

Future research activities should include longitudinal studies to disentangle the mechanisms behind the association between stigmatization and psychological maladjustment. Moreover, as the factors included in this study explained $<30\%$ of the variability in child adjustment, it will be important to examine other possible predictors, like the characteristics of family communication, child personality, or coping strategies. Notably, the question arises whether social support and good social skills might act as protective factors against the potential challenges of stigmatization. Finally, as generic measures may lack sensitivity for specific difficulties, condition-specific measures could be vital to studying the impact of facial disfigurement (Edwards et al., 2005; Patrick et al., 2007).

Regarding the clinical assistance of children with facial differences, corrective surgery may offer psychosocial benefits (Horlock, Voegelin, Bradbury, Grobelaar, & Gault, 2005). But because a complete resolution of a facial difference is rarely obtainable, medical care should be accompanied by psychological assistance. Early identification of stigma experiences and appropriate support might be crucial to enhance psychosocial adjustment and quality of life among young people with facial differences. A combination of cognitive behavioral therapy and social skills training, with inclusion of the parents, (Kish & Lansdown, 2000) could be a particularly promising approach to assisting children and adolescents with visible facial differences.

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